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Case report

Bilateral extracorporeal testicular ectopia: An extremely rare congenital anomaly



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KEYWORDS

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Abstract

Bilateral extracorporeal ectopia of the testis is an extremely rare congenital anomaly of the male external genitalia. To our knowledge, no more than 15 cases have been reported in the literature so far. We present the case of an 8-hour-old neonate with bilateral extrusion of the testes through an abnormal opening in the anterior scrotal wall. Physical examination revealed protrusion of the testes, epididymis and distal spermatic cords through a congenital anterior scrotal wall defect. All other findings were normal. After exploring the scrotum, orchidopexy was performed and the scrotal wall defect closed. At 3 and 6 months follow up, the testes were normal and located within the scrotum. The immediate outcome was excellent, but the long-term results remain to be seen. The related literature was reviewed.

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Introduction

Extracorporeal testicular ectopia is very rare. It is also referred to as scrotoschisis [1]. The few cases described in the literature mostly refer to unilateral scrotoschisis. The actual etiology of the anomaly is not known, but it may be due to a congenital defect in the scrotal wall,

resulting from meconium periorchitis [2,3]. The patients usually present early and are otherwise healthy. We present the case of an 8-hour-old neonate with bilateral scrotoschisis.

Case report

An 8-hour-old neonate was brought to our special-care baby unit with bilateral extrusion of the testes through an abnormal defect on the anterior aspect of the scrotum noticed at birth. His mother's pregnancy was supervised and labor was spontaneous at full term. The baby was delivered at home without any complications, as reported

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Fig. 1 Extrusion of both testes via abnormal congenital anterior scrotal wall defect.

by the persons present at the time of delivery. There was no history suggestive of other congenital anomalies and no history of maternal febrile illness. The boy's mother is a 20-year-old housewife, P₄₊₀ A₃. On examination, the baby weighed 3.4 kg. His skin was pink. He was febrile (temperature = 38.1 °C) and tachypneic with a respiratory rate of 60c/min but vesicular breath sounds. The heart rate was 176b/min and had normal first and second heart sounds. Examination of the external genitalia revealed extrusion of the testes, the epididymis and the distal part of the spermatic cords through an anterior scrotal wall defect. The paratesticular tissues and the spermatic cords were edematous with dark yellow discoloration, especially on the medial aspect (Fig. 1). The scrotal wall defect measured about 4 cm in diameter at the penoscrotal junction (Fig. 2). The scrotal sac was well-shaped but empty. The penis was normal and the hernia orifices were intact. All other findings were normal. Laboratory



Fig. 2 Anterior Scrotal wall defect after orchidopexy.



Fig. 3 After closure of anterior scrotal wall defect and orchidopexy.

work-up and abdomino-pelvic ultrasound scan were within normal limits. The baby was given parenteral antibiotics; the testes were cleaned with normal saline solution and covered with povidone iodine-soaked gauze. Twenty-four hours later, the scrotum was explored. The testes were cleaned again with normal saline solution and orchidopexy was performed. The anterior scrotal wall defect was closed in two layers (Fig. 3). The patient was discharged home 4 days after admission. At 3 and 6 months follow up, the testes were normal and located within the scrotum (Fig. 4).



Fig. 4 Three months after treatment.

Discussion

Extracorporeal testicular ectopia is a rare congenital anomaly of the male external genitalia. It is the extrusion of the testis or testes through an abnormal congenital defect on the scrotum. It is also referred to as scrotoschisis [1], especially when the testes extrude through scrotal defects. But the term has also been used for testes extruding through an inguinal canal defect, although in this case the term bubonoschisis would be more appropriate, as suggested by Haidar et al. [4]. The incidence of this anomaly is not known, and to our knowledge no more than 15 cases have been reported in the literature so far [5]. There is no evidence of racial predisposition, but this is the third case reported from Nigeria [6,7]. Unlike intracorporeal ectopia which is relatively common and attributed to abnormalities of testicular descend, extracorporeal testicular ectopia is rare and its etiology is as yet unexplained. The most popular theory suggests meconium periorchitis [2,3,8,9] with subsequent necrosis of the scrotal wall to be the cause of extracorporeal testicular ectopia. This theory is supported by the presence of greenish or yellowish paratesticular and spermatic cord discoloration seen in our case and histological evidence of meconium residues found in some other cases [3]. The mechanism of the development of periorchitis remains controversial. While some authors suggested trickling of meconium into the patent processus vaginalis and scrotum following intrauterine bowel perforation (meconium peritonitis) [10], others suggested it to be a result of rupture of an intrauterine incarcerated inguinal hernia. Gongaware et al. [1] suggested periorchitis to be due to a failure of differentiation of scrotal mesenchyme, resulting in a scrotal wall defect with evisceration of the jellylike gubernacular bulb. Other authors proposed arthrogryposis causing mechanical compression of the scrotum by the fetal feet to be the cause of periorchitis [11]. Early amnion rupture and amnion band sequence mechanisms have also been suggested [12]. Furthermore, a few cases of iatrogenic scrotoschisis following Cesarean section have been described [13,14].

Presentation is usually early as seen in this case. This may be due to the fact that external genitalia being the organ of sexual identity are the first part of the baby to be inspected after delivery. Most of the reported cases showed a unilateral defect in an otherwise normal male neonate. Our patient presented with bilateral scrotoschisis which is extremely rare. There have been some reports on cases associated with intestinal atresia, Beckwith–Wiedemann syndrome, ruptured omphalocele [2] and meconium periorchitis [8–10]. Some patients present with associated testicular torsion [6]. Our patient did not have any associated congenital anomaly, but he displayed features of neonatal sepsis with was treated with antibiotics. Treatment of scrotoschisis included cleaning the testis, scrotal exploration, orchidopexy and closure of the scrotal wall defect. The immediate outcome is usually good.

Conclusion

Scrotoschisis is a rare congenital anomaly. Its etiology is poorly understood, however meconium periorchitis is the most favored explanation. The treatment of choice consists of cleaning the testis/testes, scrotal exploration, orchidopexy and closure of the scrotal defect and results in a good immediate outcome.

Conflict of interest

The authors have no conflict of interest to declare.

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